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Case Report

Odontogenic Keratocyst in Posterior Mandible: A Case Report

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Abstract

Odontogenic keratocyst (OKC) is a common developmental odontogenic cyst affecting the maxillofacial region that arises from the dental lamina. The OKC is distinctive among jaw cysts and has tendency toward recurrence along with aggressive clinical behavior. The recurrent rate of OKC is 25–30%. The following is an article of a case report of a 26-year-old male who presented with OKC involving right mandibular molar tooth region.

Key words: Carnoy's solution, Keratocyst, Marsupialization, Odontogenic

INTRODUCTION

Odontogenic cysts are the most common form of cystic lesions affecting the maxillofacial region. They are classified traditionally into a developmental group, including keratocysts and dentigerous cysts, and an inflammatory group, including radicular cysts.[1] The odontogenic keratocyst (OKC) was first described by Philipsen (1956), is now designated by the World Health Organization as, a keratocystic odontogenic tumor (KCOT).[2] It is defined as "a benign uni or multicystic, intraosseous tumors of odontogenic origin, with a characteristic lining of parakeratinized stratified squamous epithelium and potential for aggressive, infiltrative behaviors."[3] The percentage of OKCs versus other cysts of the jaws as given by different authors are as follows:[4] Hjorting-Hansen *et al.* (1969)[5] and Toller (1972)[6] as 11%; Brannon (1976)[7] and Payne (1972)[8] as 9%; and Pindborg and Hansen (1963)[9] as 7%. Growth is chiefly in the anteroposterior dimension, and the lesions may attain remarkable size without significantly deforming the jaw skeleton. The particular tendency to rapid growth is due to the higher activity of the epithelial cells of the cyst lining, stimulating osteolytic activity of prostaglandin substances in the cell population of the cyst lining, and the higher accumulation of hyperkeratotic scales in the lumen of the cyst, resulting in greater difference in hydrostatic pressure.[10]

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CASE REPORT

A 26-year-old male patient reported to the Department of Oral and Maxillofacial Surgery, Guru Gobind Singh College of Dental Sciences and Research Centre, Burhanpur (Madhya Pradesh), with the chief complaint of swelling in lower right side of face for the past 4 months. There was no history of any trauma or discharge. A medical history revealed that there was no systemic illness present. There was no history of any deleterious habit such as smoking, tobacco or betel nut chewing, and alcohol, when asked for personal history.

The extraoral examination of the patient revealed diffuse swelling on lower right side of face, that is, approx. 2 cm × 3 cm in size extending anteriorly from right parasymphysis region to the angle of the mandible posteriorly, superiorly, it extended from ala tragus line and inferiorly up to lower border of mandible [Figure 1]. On palpation, it was hard

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in consistency, non-tender, non-fluctuant, and a febrile to touch.

Intraoral examination revealed that there was obliteration of buccal vestibule wrt. 45, 46, and 47 teeth region, surrounding mucosa was reddish in color [Figure 2]. On palpation, it was hard in consistency, non-tender and no discharge was present. On aspiration, straw-colored fluid was present. With the above clinical finding, provisional diagnosis of OKC wrt. 45, 46, and 47 tooth region with differential diagnosis of unicystic ameloblastoma was made.

The panoramic radiograph (orthopantomogram) showed a well-defined unilocular radiolucency wrt. 44 to 48 tooth region with well-demarcated borders and thinning of cortical bone along with cortical expansion in body region. Furthermore, impacted 45 with resorption of root was noted wrt 46 and 47 [Figure 3]. Radiographic diagnosis of OKC was made along with differential diagnosis of unicystic ameloblastoma and dentigerous cyst. The panoramic radiograph (OPG) showed formation of bone over the inferior border of mandible [Figure 4]. After taking the informed consent of the patient, marsupialization was done under local anesthesia. Under local anesthesia, through intraoral incision, the full-thickness mucoperiosteal flap was raised. Enucleation of the lesion and removal of the involved teeth were accomplished [Figures 5 and 6]. Then, peripheral ostectomy of the whole surgical bed was completed followed by a single application of Carnoy's solution. The thinned out inner cortical lining of the bone was removed. The cheesy material and cystic lesion were sent for histopathologic examination [Figures 7 and 8]. Finally, the wound closure was done with 3-0 mursilk suture [Figure 9].

Figure 1: Extraoral swelling extending anteroposteriorly from corner of mouth to angle of mandible and superoinferiorly from tragus to inferior border of mandible

Figure 2: Intraoral swelling w.r.t. 44–46 tooth region

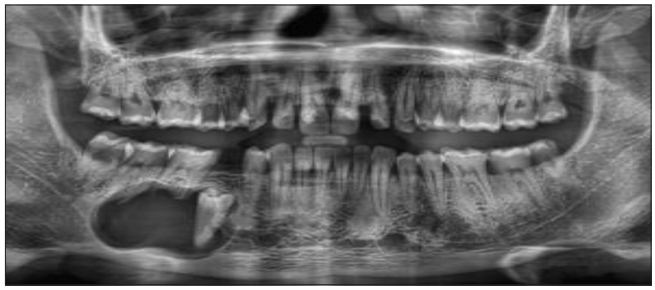

Figure 3: Panoramic radiograph showing unilocular radiolucency along with impacted 45

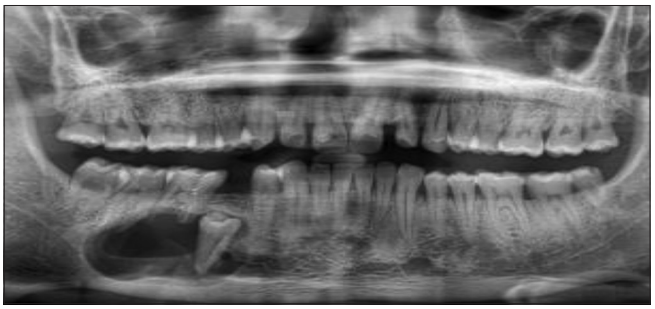

Figure 4: Panoramic radiograph showing formation of bone over the inferior border post marsupialization

Figure 5: Keratin removal

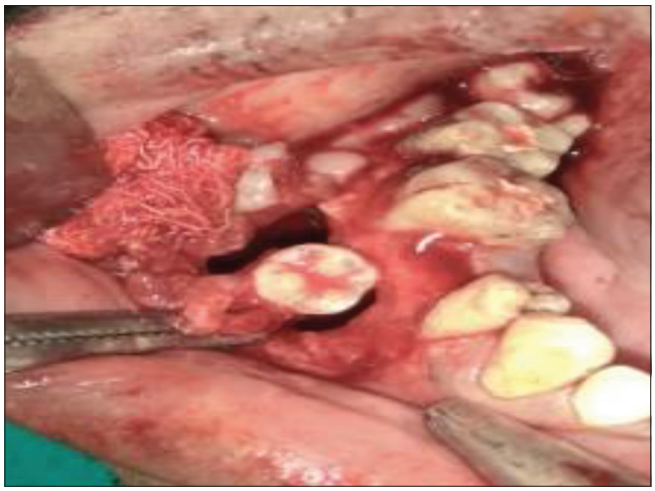

Figure 6: Removal of impacted tooth

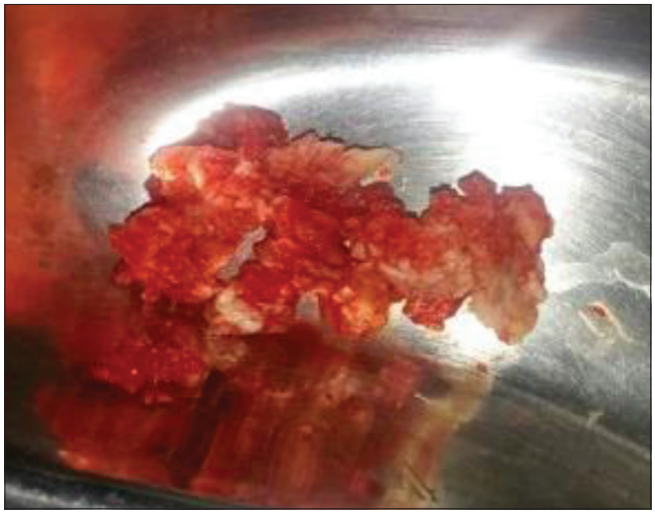

Figure 7: Complete removal of keratin

Histopathological examination of specimen revealed a cystic lining with orthokeratin layer, stratified squamous cells of 6–8 cell thickness, no daughter cyst, or epithelial detachment was found. The basal layer was composed of palisaded layer of cuboidal cells with hyperchromatic nucleus. The overall features were suggestive of OKC. Henceforth, the final diagnosis of OKC wrt. 45, 46, and 47 tooth region was made.

DISCUSSION

The OKC is a unique and prevalent clinical and histologic lesion with aggressive nature. It usually arises in the dental lamina, but some suggest a probable origin from basal cell component.[11] About 70% or more cases involve the mandible, especially in the third molar, angle, and ramus areas. Next, most common site of occurrence is the maxillary third molar followed by mandibular premolar and maxillary canine region.[12] There are few factors which led to recharacterization of the keratocyst as KCOT.[3] The KCOT

Figure 8: Cystic lining post-enucleation

Figure 9: Suturing and wound closure

exhibits locally destructive and highly recurrent behavior and is characterized by parakeratinized epithelium, in contrast to the orthokeratinized variant seen in OKC. KCOT reveals budding of the basal layer into the connective tissue and frequent mitotic figures. Furthermore, they are associated with inactivation of PTCH, the tumor suppressor gene.

Radiographically, OKC presents predominantly as a unilocular radiolucency with well-developed sclerotic borders. They may also present as a multilocular radiolucency with a ratio of unilocular to multilocular varying from 3:112 to 1:1.3. Multiple keratocysts are frequently associated with the bifid rib basal cell nevus syndrome (Gorlin syndrome).[13] Differential diagnosis includes dentigerous cyst (in OKC, the cyst is connected to the tooth at a point apical to cementoenamel junction), ameloblastoma (usually multilocular, no straw-colored

fluid on aspiration), traumatic cyst (unilocular with scalloped margins, rarely show cortical expansion), giant cell granuloma (usually in anterior region of jaw), and odontogenic myxoma.[14]

Treatment modalities for OKC include a conservative approach that is marsupialization and decompression which lead to ultimate complete resolution of the cystic lesion. They are preferred to preserve bone, teeth, and preventing damage to other vital structures, also for decreasing the chances of pathologic fracture. The principle behind these procedures is to decrease the cystic osmotic pressure by exposing it to the surrounding oral cavity. It is helpful in a bone deposition at the periphery of the lesion and also a progressive reduction in the cyst size.[15] Other treatment modalities are extensive surgeries, *en bloc*, and segmental resection.[16]

Two-step procedure in the management of OKCs is decompression which involves the placement of a surgical drainage tube, the following enucleation after the cyst has reduced to a manageable size. Enucleation is the complete and intact removal of a cystic lesion by surgically husking it from the surrounding tissues. About 17–56% of the recurrence rate of enucleation has been reported. Because of this, many surgeons prefer a combination of enucleation and adjunctive therapies to eliminate any residual cyst lining and islands within the cyst wall. Adjunctive therapy includes the application of Carnoy's solution which destroys cyst remnants using chemical cautery, intended to reduce recurrence rates.[17,18] It is often used as a complementary treatment of lesions such as the KCOT and is composed of 3 ml of chloroform, 6 ml of absolute ethanol, 1 ml of glacial acetic acid, and 1 g of ferric chloride.[18]

OKC has a particular tendency to recur after surgical treatment. The first to point out this peculiarly aggressive behavior was Pindborg and Hansen (1963).[9] Recurrence is encountered more often in mandibular OKC, particularly those in the posterior body and ascending ramus.[17] A total of 18% reduction in the recurrence potential is noted, when both combination of enucleation with adjunctive treatment are performed.[19]

CONCLUSION

KCOT has been identified as a "tumor" after observation of its biological behavior and genetic abnormalities consistent with neoplastic progression. This case report of OKC was noticed in a 26-year-old male patient on the right side of mandible showing the clinical and radiographic presentation, diagnosis, treatment, and follow-up. Research is still going on appropriate treatment modalities for OKCs because of it is genetic and molecular basis of pathogenesis. Surgeons should thoroughly examine each case individually and should provide better treatment options to the patients.

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